Case Report

Juvenile recurrent parotitis: Report of two cases

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Abstract

Juvenile recurrent parotitis is a recurrent, nonobstructive, and nonsuppurative inflammatory condition of the parotid gland of uncertain etiology in young children. It is characterized by multiple episodes of unilateral or bilateral parotid inflammation over a period of years. Two cases of juvenile recurrent parotitis in a 3-year-old girl and a 6-year-old boy are reported. Both presented with multiple episodes of the parotid gland swelling. Investigations for infective etiology, an autoimmune disorder, Sjogren's syndrome, and immunodeficiency were negative. Diagnosis was established clinically and confirmed by ultrasound. They were managed symptomatically and recovered in 6-7 days. The parents were counseled about the benign nature of the condition and its resolution by puberty.

Key words: *Children, Parotitis, Recurrent*

Juvenile recurrent parotitis is an uncommon recurrent, nonobstructive, and nonsuppurative inflammatory condition of the parotid gland of uncertain etiology [1]. It is seen in young children and characterized by multiple episodes of unilateral or bilateral parotid inflammation over a period of years [2,3]. Very few cases are reported till date. Two cases of juvenile recurrent parotitis in a 3-year-old girl with bilateral involvement and a 6-year-old boy with unilateral parotid involvement are reported.

CASE REPORTS

Case 1

A 3-year-old girl presented with painful swelling of both the parotid glands. There was no history of fever or respiratory symptoms. She was immunized for age and had received MMR and Hepatitis B vaccine. Examination of the oral cavity revealed normal throat with no dental caries. There was no redness at the opening of Stenson's duct. She had seven such episodes for the past one year with each episode lasting for 6-7 days and received analgesics and oral antibiotics during three episodes. There was no history of dryness of mouth and eyes, joint pains and swellings, and skin rashes. There was no family history of similar complaints. She was a child with normal growth, local examination revealing smooth, nontender parotid gland, soft to firm in consistency. There was no erythema around Stenson's duct opening. Systemic examination was normal.

Investigations revealed normal hemoglobin, total leukocyte count, and erythrocyte sedimentation rate (ESR) (8 mm at end of 1 h). Ultrasound revealed bilateral enlarged parotid glands with multiple subcentimeter (4-5 mm) hypoechoic areas suggesting sialectasis. Other salivary glands were normal. Serum immunoglobulin (Ig) levels were normal; IgA - 1.10 g/L (normal 0.17-2.90 g/L), IgG - 12.70g/L (normal 4.0-15.9 g/L) and IgM - 1.45g/L (normal 0.34-3.48 g/L). Human immunodeficiency virus (HIV) serology was negative. Serum anti-nuclear antibody, dsDNA and rheumatoid factor along with antibodies to nuclear antigen Ro (SSA) and La (SSB) were

negative, and Schirmers' test for Sjogren's syndrome was noncontributory.

Case 2

A 6-year-old boy presented with the fourth episode of right parotid gland swelling. He had associated mild fever and had been immunized with MMR and Hepatitis B vaccine. There was no history suggestive of an autoimmune disorder. His growth and systemic examination was normal. Local examination revealed enlarged firm right parotid gland without any erythema. Oral cavity was normal, and there was no pus discharge from the parotid duct on bimanual palpation. He was managed symptomatically and had received oral antibiotics for the same during last three episode. Complete blood count was normal, HIV test was negative. IgA levels were normal. Ultrasonography (USG) of the right parotid gland revealed findings suggestive of sialectasis. In addition, there was no calculus, abscess, or a mass lesion demonstrated on sonography. The left parotid and other salivary glands were normal.

Considering a diagnosis of juvenile recurrent parotitis, both these children were treated with analgesics and advised to take plenty of oral fluids. They were asymptomatic within 5-7 days. The parents were counseled regarding the benign nature of this condition, and they are being followed up regularly.

DISCUSSION

Juvenile recurrent parotitis is an uncommon disorder of childhood characterized by repeated episodes of swelling and pain in the parotid gland. The disease is more common in males and usually manifests between 3 and 6 years of age [1,2]. Biphasic age of distribution at 2-5 years and 10 years has been reported by Leerdam et al. [3] Parotid involvement is mostly unilateral but can be bilateral [3-5]. In the reported cases, there was bilateral involvement in the girl and the boy had unilateral involvement. Children usually present with swelling, pain, and fever. Symptoms usually last for 2-7 days, with the mean frequency of eight episodes/year [4]. The girl had seven episodes and the boy had four episodes in the previous year and the symptoms lasted for 5-7 days in both the cases.

The etiology and pathogenesis of juvenile recurrent parotitis of childhood remain uncertain. Various factors that have been suggested include congenital ductal malformations, hereditary genetic factors, viral or bacterial infections, allergy, and local manifestation of an autoimmune disease [1]. The reported cases had normal blood counts ruling out infectious etiology. They had normal ESR. Rheumatoid factor, anti-nuclear antibodies, and antibodies to nuclear Ag SS-A (anti Ro) and SS-B (anti La) done in a girl child were negative, thus ruling out Sjogren's syndrome. Recurrent parotitis may be the first manifestations of HIV infection or immune deficiency disease [6]. Both the children were screened for HIV, which was negative and had normal IgA levels thus the possibility of HIV infection or immune deficiency and IgA deficiency was excluded.

The diagnosis of juvenile recurrent parotitis is usually made on a clinical basis as suggested by detailed history taking and adequate physical examination. It is distinguished from suppurative parotitis by the inability to express the pus from the parotid duct. USG of the parotid glands should be done to confirm the diagnosis although parotid sialogram is a hallmark in the diagnosis of juvenile recurrent parotitis. As sialogram is an invasive procedure, ultrasound is now the preferred investigation and found to be superior [7]. The typical features of recurrent parotitis are the formation of punctate or globular sialectasis scattered throughout the gland without any stones or destructive changes. USG also provides extra information such as the presence of stones (sialoliths), abscesses, or mass lesions. In the reported cases, USG revealed features suggestive of sialectasis.

Computed tomography and magnetic resonance (MR) sialography are also the other investigations for establishing the diagnosis [8,9]. MR sialography is a recent noninvasive 3D imaging technique that allows the imaging of the salivary ducts. It accurately depicts findings such as sialectasis and signal intensity changes in the gland depending upon the phase of the disease (acute vs. chronic inflammation).

Juvenile recurrent parotitis is usually a self-limiting condition. In 80-90% of the patients, the symptoms spontaneously resolve by puberty [2,10]. The recurrent attacks are treated conservatively with

analgesics. Role of antibiotics is controversial [3,10]. However, no prophylactic therapy is available. In addition, attention to good oral hygiene, massage of the parotid gland, warmth, use of sialogogic agents like lemon juice may be helpful in reducing the attack frequency. A new treatment modality of irrigation and dilation under direct vision by endoscopically guided miniature surgical instrument has been found to be successful in preventing the recurrence [1]. Reassurance of the family is needed about the benign course of the disease as most of the cases resolve by puberty. However, the child should be screened for Sjogren's syndrome and immunodeficiency and kept under regular follow-up.

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